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CASE REPORT

Oral glial choristoma

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Summary Heterotopic brain tissue is considered to be one of the very rare choristomatous lesions involving the oral cavity. This report describes the morphologic and immunohistochemical features of one case of glial choristoma arising in the tongue and discuss its probable embryogenesis.

Introduction

Choristoma is a general term referring to normal tissue found in an abnormal location. Brain heterotopias are generally considered congenital malformations, embryologically related to encephaloceles from which they can be differentiated by the absence of an anatomic connection with the brain.[1] Heterotopic brain tissue masses are unusual and are generally located in the head and neck region. Most of the cases reported are in the nasal region, with or without connection to the brain, and are referred as nasal gliomas. Oral glial choristomas are extremely rare lesions usually affecting neonates and do not show intracranial communication.[2] The main affected site is the palatopharyngeal complex area, followed by the tongue. The purpose of this article is to report a case of glial choristoma of the tongue.

Case report

A 28-year-old man was referred to the Oral Diagnosis Service, Pontifícia Universidade Católica de

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Minas Gerais, for evaluation of tumoral lesion at the dorsum surface of the tongue present for the last 10 months. Physical exam revealed that the lesion was symptomatic, with approximately 2 cm × 2 cm diameter, showed a firm consistency and an intact overlying mucosa (Fig. 1A). The differential diagnosis included granular cell tumor, lingual thyroid, neurofibroma, and neurilemoma. An excisional biopsy was performed. During the surgery, a cystic cavity with serous fluid was noted.

Microscopic examination showed a well-defined circumscribed submucosal mass consisting of a proliferation of mature neuroglial tissue and scattered neural axons (Fig. 1B). The glial tissues was formed by cells with one or more nucleus, basophilic fibrillar cytoplasm resembling astrocytes together with rows or clusters of oligodendroglial formed by small dark staining cells with characteristic clear perinuclear halos (Fig. 1C). Microglial-like cells with small, dark, rodshaped nuclei were ubiquitously found (Fig. 1D). The glial cells were supported by a fibrillar homogenous eosinophilic stroma and no choroid plexus was found. Cystic area formed by degeneration of the neoplasia was observed.

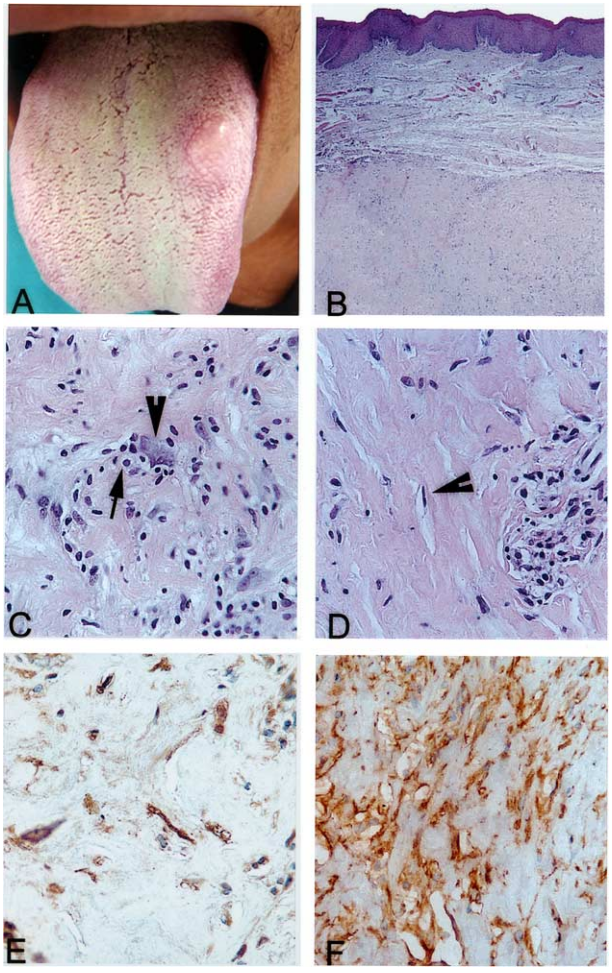

Figure 1 (A) Clinical appearance of the lesion. (B) Low microscopic view of a well-defined circumscribed submucosal mass of neuroglial tissue (HE, original magnification ×100). (C) High microscopic view showing neuroglial tissue formed by cells resembling astrocyte (arrowhead) together with rows or cluster of oligodendroglial with characteristic perinuclear halos (arrow). (D) Microglial-like cells with elongated nuclei (arrow) (HE, original magnification ×400). The glial tissue was strongly positive for S-100 (E) and GFAP (F) proteins (original magnification ×400).

Paraffin-embedded tissue blocks of the tumor were cut at 3 μm and subjected to the biotin–streptavidin amplified system for the immunolocalization of glial fibrillar acidic protein (GFAP) (Clone 6F2, diluted 1: 100) (Dako Corporation), S-100 (Clone Z0311, diluted 1: 200) (Dako Corporation), neuron-specific enolase protein (NSE) (Clone A0587, diluted 1: 100) (Dako Corporation), muscle actin (Clone HHF-35, diluted 1: 100) (Dako Corporation), and vimentin (Clone V9, diluted 1: 100) (Dako Corporation). Antigen retrieval was performed to GFAP, NSA and vimentin with citrate buffer (0.01 M, pH 6.9 in steamer/95 °C, 30 min).

The results demonstrated that the glial tissue was strongly positive for S-100 and GFAP proteins (Fig. 1E and F). Scattered positive cells to NSE and vimentin proteins were observed together with negative staining to muscle actin. A diagnosis of glial choristoma was made. The patient has been follow-up and did not present recurrence after 6 months.

Discussion

Heterotopic brain tissue is a rare entity frequently located in nasal region. Although brain tissue or tumors do not occur usually in the oral cavity, oral pathologists must be familiarized with their histological features. Oral glial choristoma is generally reported in the palatopharyngeal complex area of infants and children without gender and ethnic susceptibilities.[2] The tongue is the second most frequently reported site. While lesions in the tongue never communicate with the brain, communication may occur when the primary mass locates in the palatopharyngeal area. This fact is even more frequently observed with nasal gliomas.

Histologically, diverse histopathologic features are noted in most of the cases of oral glial choristoma. Some reports mention only the presence of neuroglial tissue without further description. Pure neuroglial tissue intermixed with neuron cells, with

or without choroids plexus and clefts lined by ependymal-type epithelium are described.[3–5] In the present case neuron axons intermixed with cells resembling astrocytes, oligodendroglial and microglial cells were observed. The immunohistochemical profile in the present case was according to the literature.[4,6] The neuroglial tissue is intensely positive for GFAP and S-100 and weakly positive for NES and vimentin.

The embryological disturbance related to the pathogenesis of glial chroristoma located in the tongue seems to be different from brain heterotopias in other sites.[1] The nasal and palatopharyngeal lesions may arise from herniation of neural tissues through an arrested closure in the osseous cranium or by an initial overgrowth of the developing neural tube preventing closure of the cranial opening. On the other hand, the glial choristoma of the tongue may have a different pathogenesis. According to the current theory, the tongue musculature is derived embryologically from occipital myotomes that differentiate into myofibroblasts that migrate to the stomatodeum. Lingual heterotopic glial tissue masses might develop from a nest of pleuripotent cells, which become separated before complete fusion of the neural tube, and integrate within those migrating myoblasts to finally reach the tongue.[2]

The complications of oral glial choristoma depend of the site of occurrence and size of the lesion. While large lesions at the palatopharingeal area may be life-threatening due to airway obstruction, most of the lesions of the tongue did not cause any complication. A previous report of a deeply infiltrative lesion located in the base of tongue was followed by a history of dyspnea, cyanosis, bradycardia, and dysphagia.[7] Conservative excision is adequate for glial choristoma and the rare cases of recurrence are due to incomplete excision.[8]

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References

1. García-Prats MD, Rodríguez-Peralto JL, Carrillo R. Glial choristoma of the tongue: Report of a Case. *J Oral Maxillofac Surg* 1994;**52**:977–80.
2. Abdelsayed RA, Wetherington RW, Bent JP, Sharpe DE. Glial choristoma of the tongue. A case report and review of the literature. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1999;**87**:215–22.
3. Bychkov V, Gatti WM, Fresco R. Tumor of the tongue containing heterotopic brain tissue. *Oral Surg Oral Med Oral Pathol* 1988;**66**:71–3.
4. Landini G, Kitano M, Urago A, Sugihara K, Yamashita S. Heterotopic central neural tissue of the tongue. *Int J Oral Maxillofac Surg* 1990;**19**:334–6.
5. Morita N, Harada M, Sakamoto T. Congenital tumors of heterotopic central nervous system tissue in the oral cavity. Report of two cases. *J Oral Maxillofac Surg* 1993;**51**:1030–3.
6. Ide F, Shimoyama T, Horie N. Glial choristoma in the oral cavity: histopathologic and immunohistochemical features. *J Oral Pathol Med* 1997;**26**:147–50.
7. Strome SE, McClatchey K, Kileny PR, Koopmann Jr CF. Neonatal choristoma of the tongue containing glial tissue: diagnosis and surgical considerations. *Int J Pediatr Otorhinolaryngol* 1995;**33**:265–73.
8. Ofodile FA, Aghadiuno PU, Oyemade O, Adebonojo T. Heterotopic brain in the tongue. *Plast Reconstr Surg* 1982;**69**:120–4.